Developing a sleep hygiene education intervention for children with developmental disabilities and sleep problems.

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Key points

Sleep hygiene education for behavioural sleep problems in children with developmental disabilities is a usual first-line intervention but has a limited evidence base.

A systemically developed sleep hygiene education tool for children with developmental disabilities suggests 45 different advice points for practitioners to select from.

An underpinning programme theory explains how sleep hygiene education is supposed to work to improve children's sleep.

Experience based co-design is a participatory research method which can promote professional and public involvement in service improvement.

Sleep problems in children with developmental disabilities need to be reprioritised by policy makers and awareness raised of available sleep support.

Abstract

This co-design study develops a sleep hygiene education (SHE) tool for children with developmental disabilities (DD) and behavioural sleep problems. This is underpinned by a programme theory which explains how SHE is supposed to work to improve children's sleep. In three co-design workshops, eight parents and six practitioners debated a preliminary SHE tool developed from an earlier evidence review and discussion themes generated from an earlier exploratory study into parent and practitioner experiences of SHE. This participatory research established stakeholder acceptability of the SHE tool and confirmed the often hidden contextual factors which can help or hinder SHE intervention success which informed the underpinning programme theory.

Key words/search terms

Children, developmental disabilities, sleep problems and sleep hygiene education.

Background, aim and literature review

Severe and chronic sleep problems such as difficulties falling asleep and multiple night awakenings are common in children with developmental disabilities (DD) (Bonuck et al, 2012). Such sleep problems are linked to negative outcomes for the child and include impaired concentration, mood

and behavioural difficulties (Beresford et al, 2012, Mazurek et al, 2015). Furthermore, family members are known to experience increased stress, relationship and employment difficulties as a result of managing children's sleep difficulties on a long term basis (Bourke-Taylor et al, 2013, Roberts et al, 2019).

Practitioners are advised to firstly identify and address physical causes of sleep problems such as epilepsy or breathing difficulties and assess for behavioural causes linked to parental management style such as inconsistent boundaries (Malow et al, 2013). Usual first-line treatment for sleep problems that have behavioural origins is sleep hygiene education (SHE) (Blackmer et al. 2016) which advises parents on creating positive sleep environments and routines that promote optimal sleep.

In a 2013 scoping review of SHE for sleep problems in children with DD (Sutton, 2017) six different categories of SHE advice components were identified supported by mixed evidence: sleep timing, bedtime routines, behaviour management, environment, physiological factors and communication adaptations. In addition, limited evidence was found supporting SHE as a credible, primary sleep intervention. Just two intervention studies explored SHE as a stand-alone treatment: Adkins et al (2012) researched outcomes on children with autism and reported improvements in sleep efficiency, whereas Piazza, Fisher & Sherer (1997) concluded SHE was less effective than a comparative behavioural intervention however, the study lacked methodological rigour.

One qualitative study investigated professional and parent views of SHE combined with other behavioural interventions (Beresford et al, 2012) and identified enablers to intervention success such as practitioner's specialist sleep knowledge and barriers such as parent's lack of readiness to engage. However, this study's findings had restricted applicability to SHE as a distinct intervention. In addition, the scoping review found no evidence of how policy or organisational factors could affect intervention success. Overall, review findings highlighted a need for further research into SHE and its application to sleep problems in children with DD.

To further understanding, the complexity involved in delivering SHE to families was considered. It was acknowledged that many factors such as social context, parent/practitioner relationships and levels of support could impact on the success or failure of SHE in improving children's sleep. The importance of identifying these often hidden elements is essential according to Funnell et al (2011) as practitioners need to be aware of how complex interventions like SHE create change, to ensure only best practice is repeated. This can be achieved by developing a programme theory underpinning the intervention which helps to explain the process of change in a detailed way (Chen, 2015). Furthermore, in today's challenging economic climate, there is a renewed urgency to ensure practitioners explicitly understand the SHE advice they give and only deliver care which is relevant to individual need (Bradley et al, 2014).

The Medical Research Council (MRC) (2000, 2008) offers a helpful framework to develop complex interventions such as SHE and underpinning evidence. The process begins with an evidence review (Sutton, 2017) which informs additional exploratory research with stakeholders to build intervention understanding. Hence, a qualitative study investigating the SHE experiences of nine parents of children with DD and practitioners from health and social care (Sutton et al, 2019) was undertaken. Study findings were synthesised with review findings and summarised into six overarching themes (Table 1) which told the overall story of the evidence base, stakeholder views on SHE and how it should be implemented. This informed a co-design study (Sutton et al, 2018) which aimed to:

- Systematically develop and confirm a SHE tool for children with DD and
- Develop a programme theory underpinning SHE to offer an explicit understanding about what SHE does, how it is delivered and how it is supposed to work to improve sleep.

Method-

Research design.

Following a participatory research methodology (Creswell, 2007), experience based co-design (EBCD) (Point of Care Foundation, 2018) was adopted as a participatory research method which champions public and professional involvement in planning improvements in care. The EBCD method recommends numerous research stages. The preliminary stages mirrored the review and exploratory studies which produced six explanatory themes. The interim stages involved the illustration of these themes via an auditory podcast derived from exploratory study parent interviews (Sutton, 2019). In the final EBCD stages the podcast was aired to trigger debate in three co-design events with co-designers: eight parents of children with DD and sleep problems and six practitioners with experience of SHE recruited from social care and voluntary organisations. Each event ran for two hours and was held at parent-friendly voluntary organisation meeting rooms. Co-designers were invited to deliberate each discussion theme and:

- Establish how parents and practitioners could work together to ensure SHE was effective
- Describe an exemplar parent journey in securing professional SHE
- Develop understanding of what makes SHE work
- Confirm the acceptability of the developing SHE tool.

Any new insights or emerging ideas were captured on a wall-sized visual representation based on a person centred planning tool: The Planning Alternative Futures with Hope (PATH) model (Sanderson et al, 2012) which helped to consolidate findings and keep discussions focused (Figure 1). Co-design events were audio taped and video recorded then transcribed verbatim. Data were thematically analysed (Braun et al, 2006) in relation to the discussion themes (Table 1). Evidence review, exploratory and co-design study findings were then synthesized to iteratively construct a SHE tool for children with DD and underpinning programme theory.

Findings

SHE Tool

A SHE tool was developed which incorporated six categories and 45 advice components for practitioners to select from (Table 2). Advice points were only included if they were evidenced by a minimum of two review citations or one citation plus further research evidence or co-designer agreement to ensure the tool was valid. Advice for practitioners on the correct use of the SHE tool is provided in the supporting programme theory.

Programme theory

The six discussion themes (Table 1) were reframed as SHE intervention desired outcomes, and the activities and processes, resources, programme and non-programme factors (hidden contextual factors) that contribute to the achievement or non-achievement of each desired outcome were listed in a matrix (Figure 2).

SHE desired outcome 1- Parents and practitioners have a shared understanding of what a sleep problem is.

This pre-implementation outcome of SHE describes how a common appreciation should be developed between parents and practitioners of what constitutes a sleep problem in a child with DD. This is facilitated by training generic and sleep practitioners in sleep problem recognition and encouraging practitioners to adopt a positive attitude toward sleep; raising parents' expectations that children's sleep can improve. Contextual factors that may prevent parents from recognising children's sleep problem include adopting coping strategies such as co-sleeping which can mask the

problem or other family members insisting on a stoic attitude towards managing their child's sleep. The SHE intervention also aims to screen children with DD for sleep problems through screening all children as standard practice. Parent co-designers felt this would help to 'normalise' inquiries about sleep problems influencing their receptiveness to admitting sleep problems. In addition, parents wanted to be empowered to independently screen for children's sleep problems and requested the development of screening tools embedded in their personal child health record. Lastly, the intervention works to promote a public conversation about children's sleep problems through media campaigns to promote more parents seeking early sleep help. This leads to the following outcome.

SHE desired outcome 2- Sleep services are well publicised and accessible for parents.
This pre-implementation outcome explains how available sleep services should be actively publicised and easily accessible for parents. This is achieved by ensuring sleep services for children with DD accept direct referrals from parents, operate short waiting lists, offer good geographical coverage and broad referral criteria which does not exclude children based on the child's age or type of DD. Parent co-designers also felt it was important they could 'normalise' their child's sleep problem by accessing mainstream sleep services first, who could signpost them to specialist sleep services for children with DD afterwards. This was identified as significant in bringing about the change of parents deciding to access the sleep help that they need. Finally, the intervention works to ensure specialist sleep services are widely publicised to raise awareness amongst parents and generic practitioners. However, the contextual factor of parents having literacy issues or limited internet access could prevent them from learning about available support. This leads to the next two intermediate outcomes which can be addressed simultaneously.

SHE desired outcome 3- Parents and practitioners develop a safe and supportive relationship.
This intermediate outcome describes the need for parents and practitioners to build a trusting working relationship before parents feel able to follow SHE advice. The intervention works to ensure practitioners demonstrate good interpersonal skills when working with parents which is achieved through appropriate training. Parent co-designers voiced the importance of practitioners appreciating their situation, communicating sensitively, offering reassurance which helps create confidence in the practitioner's ability to give sound advice. However, the contextual factor of some parents being mistrustful of practitioners due to past experiences was acknowledged as a potential barrier to intervention success. The amount and type of support offered to parents throughout their contact with sleeps services was also highlighted as significant. For example, parents wanted the option of home visits as this was where they felt most comfortable discussing the sensitive topic of sleep. The quantity of support also needed to be driven by parents' need. The development of a supportive partnership can take time and can be built whilst the sleep problem is being assessed in the following outcome.

SHE desired outcome 4- Parents and practitioners improve their knowledge of the sleep problem.
This intermediate, direct focus of SHE effort describes the need for parents and practitioners to increase their knowledge of the nature and causes of the sleep problem. The intervention works to provide a comprehensive sleep assessment process through adequate resourcing of the practitioners' time to observe the child at home at bedtime, over multiple sessions whilst liaising with other members of the multi-disciplinary team and partner organisations. Co-designers felt this helps parents to feel practitioners have invested enough time getting to know their child and will be basing their advice on the information collected. Various contextual factors were identified that could affect success such as whether parents gave assessment information honestly and if practitioners had an effective working relationship with partner organisations. The intervention also works to provide a competent sleep assessment which involves firstly screening the child for physical and psychological co-morbidities which may be causing the sleep problem. If co-occurring conditions are overlooked this can affect the success of the SHE intervention. Sleep training also equips

practitioners with the skills to utilise sleep histories, sleep diaries and validated outcome measures to uncover multiple causal factors, understand the nature and establish a baseline recording of the sleep problem. Practitioners will also be skilled at interpreting sleep assessment findings in partnership with parents and using a psychological formulation to summarise the child's strengths and behavioural causes of sleep problems. This may include identifying sleep disorders such narcolepsy or circadian rhythm sleep wake disorders which require alternative interventions to SHE. Non-programme factors which could hinder success and may be largely outside the practitioner's control include the willingness of parents to fully engage in the sleep assessment process which it is acknowledged requires significant commitment. The importance of providing a supportive sleep assessment process was therefore very important which included suggesting alternative methods of recording sleep information using visual prompts, easy read sleep diaries or phone apps. In summary, achieving a detailed knowledge of the sleep problem is a crucial step towards tailoring SHE advice and identifying the support needs of families which feeds into the following ultimate outcome of SHE.

SHE desired outcome 5- Regularity and quality of the child's sleep problem improves.

This direct focus of SHE effort describes how the child's sleep can be improved. The intervention works to deliver effective SHE through training practitioners to customise sleep advice selected from the SHE tool according to the assessment findings and needs of the family. (i.e. it is important parents are not asked to achieve all 45 advice points as this would be unrealistic and inappropriate). This may also include making appropriate referrals to other generic practitioners or support organisations based on assessed needs. Co-designers stressed the importance of backing up SHE advice with rationale and psycho-education to help motivate parents to follow it. Numerous contextual factors were acknowledged that could impact on intervention success including whether parents had mental health issues or the child had competing health issues. This makes explicit the complexity of SHE.

The intervention also works to ensure practitioners support parents in following SHE advice through resourcing their time to offer the required level of support. Within this was the need to make an active offer of parents bringing peer supporters to all appointments to address any perceived imbalances of power. Finally, resourcing of paid 'parent buddies' within sleep teams was recommended by co-designers to coach parents in following advice. At the boundary of the programme is also the contextual factor of whether partner organisations such as schools accurately follow sleep advice such as not allowing daytime sleep, or jeopardise progress made by parents by failing to do so. This continues to demonstrate the complexity of support needed by parents to follow SHE advice. When the regularity and quality of the child's sleep is improved, quality of life may then improve for the child and family which the next outcome represents.

SHE desired outcome 6- Quality of life improves for the child and family.

This ultimate outcome describes how quality of life can be improved as bi-product of successful SHE implementation. SHE works to measure improvements through running quality of life outcome measures at the start and end of the intervention. Alternatively, qualitative evaluation questionnaires may be administered as a follow up to the intervention to capture how parents feel their quality of life has been improved. The completion and chasing up of this documentation requires resourcing of practitioner's time and additional commitment from practitioners. Whether parents have the time and motivation to complete the documentation is also acknowledged as an external factor that could impact on the success of this outcome. The intervention also works to help parents feel supported to maintain sleep progress made by offering follow up support for families. Co-designers highlighted it was important parents felt reassured by the offer of continued practitioner support if needed to help sustain sleep improvements in their child. Finally, SHE works to support parents by ensuring parent support groups are available and backed by practitioners. Ongoing encouragement that can be offered by parents in a similar situation can be a powerful tool

to help parents continue with SHE advice and avoid returning to previously unhelpful coping strategies. However, it is acknowledged practitioners need to be resourced to support these groups and some parents may not be comfortable accessing these groups.

Discussion

Study findings provide a systematically developed SHE tool for children with DD. All advice points have been supported by the research of others or confirmed by the co-designers in this study. This tool is underpinned by a programme theory which offers an explicit understanding about what SHE does, how it is delivered and how it is supposed to work to improve sleep.

The programme theory had a strong emphasis on raising awareness of sleep problems in children with DD and empowering parents to ask for professional help. This concords with the ~~recent~~ Tired all the Time report (Family Fund, 2013) which argues that sleep problems need to be reprioritised by policy makers. Furthermore, the recommendation of informing parents of the rationale underpinning SHE and psycho-education is supported Beresford et al (2016) who also reported that enhancing parents sleep knowledge was influential in improving children's sleep. The recommendation that parents should continue to be supported post-intervention is also espoused in the findings of a review of behavioural sleep interventions by Kirkpatrick et al (2019).

The EBCD method was found to be an effective participatory research method which actively encouraged professional and public involvement in the research process. The audio podcast was particularly powerful in eliciting co-designer responses and feeding back parents views to practitioners. However, there was limited guidance on how to facilitate the co-design groups and keep co-designers focused. Similar limitations were reported by Locock et al (2014) who introduced additional formal tools such as Quality Circles to ensure groups stayed on track. In the first co-design event parents often digressed to broader issues around disability diagnosis or education therefore the PATH tool was successfully introduced in the second co-design event to serve as a constant visual reminder of the events' purpose and aims.

It could also be argued that 'authentic' expression of the parents' voice could be stifled by the co-design process, if they felt unable to speak candidly in the presence of practitioners. Therefore, efforts were made to facilitate events in a supportive way to minimise the effects of psychosocial factors such as obedience, dominance and conformity on group processes. This highlights a potential limitation of the EBCD method and separate stakeholder groups can be more effective in genuine participant views (Wainwright et al, 2014).

Furthermore, other participatory approaches such as the Delphi method often involve service commissioners to ensure that service improvement ideas are based on cost/resource considerations as well as clinical expertise and service user opinion (Snape et al, 2014). As this study was only representative of parent and practitioner views, future research that incorporates consultation with commissioners and policy developers would enhance the feasibility of the SHE tool and programme theory.

Conclusion:

Study findings enhance our understanding of how SHE for children with DD and sleep problems is supposed to work to improve sleep. The SHE tool and supporting programme theory contribute to the evidence base that supports SHE as a credible, distinct and first-line intervention for behavioural sleep problems in children with DD. The next step advised by the MRC framework is further studies which focus on 'modelling process and outcomes'. This involves translating the programme theory's main desired outcomes into an operational manual to be used in conjunction with the SHE tool that

can be piloted with relevant primary care teams. This is a final development task recommended prior to a main evaluative study which would test the effectiveness of the developed SHE intervention. (3255 words).

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